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Neurectomy of C2 for the Treatment of Occipital Neuralgia: Case Report

Neurectomia de C2 para tratamento da neuralgia occipital: Relato de caso

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Abstract

Occipital neuralgia (ON) is an uncommon cause of headache, and it is characterized by a stabbing paroxysmal pain that radiates to the occipital region. The present study includes a review of the literature and a case report. The etiology of this pathology can vary from traumas, infections, compressions of nerves or vertebrae, skull base surgeries, to degenerative changes and congenital anomalies. However, most of the time, the etiology is considered idiopathic. The diagnosis is essentially clinical. However, it is crucial that other types of primary headache are excluded. The treatment for ON may be based on nerve blocks, medications or surgeries. Neurectomy of the second spinal nerve is among the surgical techniques available.

Keywords
- neurectomy
- occipital neuralgia
- headache

Resumo

A neuralgia occipital (NO) é uma causa incomum de cefaleia, caracterizada por dor paroxística, em pontada, que se irradia para a região occipital. Este estudo inclui uma revisão da literatura e o relato de um caso. A etiologia desta patologia pode variar de traumas, infecções, compressões de nervos ou vértebras, cirurgias de base de crânio, até alterações degenerativas e anomalias congênitas. Contudo, na maioria das vezes, a etiologia é considerada idiopática. O diagnóstico é essencialmente clínico. No entanto, é fundamental que sejam excluídos outros tipos de cefaleias primárias. O tratamento da NO pode se basear em bloqueios nervosos, medicamentos ou cirurgias. Entre as técnicas cirúrgicas disponíveis, encontra-se a neurectomia do segundo nervo espinhal.

Palavras-chave
- neurectomia
- neuralgia occipital
- cefaleia

Introduction

According to the International Headache Society, occipital neuralgia (ON) is defined as a paroxysmal, stabbing pain that radiates to the occipital region.[1] This pathology was described in 1821 by Beruto et al, being considered an uncommon cause of headache.[2] Most cases of ON are of idiopathic origin. However, external causes such as infections, skull base surgeries, vertebral compressions, traumas and congenital anomalies may be considered as etiology.[2] Due to the variety of signs and symptoms that may manifest in patients with ON, this pathology may be mistaken for other types of primary headache.[3] Nevertheless, the diagnosis is considered essentially clinical.[3] As options for treatment, non-ablative surgical treatment techniques may be used, among which neurostimulation of the greater occipital nerve and medullary stimulation can be

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highlighted. Finally, in the ablative surgical treatment group, radiofrequency rhizotomy, Lissauer tract injury surgery, neurectomy of the greater occipital nerve, and neurectomy of spinal nerve C2 are found.[3] The objective of the present study is to report the clinical case of a patient with ON whose treatment of choice was neurectomy of the second spinal nerve.

Case Report

A 46-year-old patient, female, married, presented with a history of occipital headache of strong intensity associated with pain in the bilateral mastoid process and with burning sensation in the occipital region for ~ 8 years. She also reported the presence of paroxysms, in which the pain was exacerbated, with the presence of a trigger point bilaterally in the occipital region. The neurological examination showed a positive Tinel sign in the occipital nerve bilaterally, but without focal alterations. The patient was initially treated with: 150 mg of pregabalin every 12 hours; 25 mg of amitriptyline every 24 hours; 40 mg of fluoxetine every 24 hours; 500 mg of dipyrone + 5 mg of promethazine + 10 mg adiphenine every 8 hours; and 60 mg of codeine every 8 hours, without substantial improvement of pain. The patient was submitted to an MRI, in which secondary causes of headache were discarded. A laboratory review revealed an erythrocyte sedimentation rate of 36 mm after 1 hour. The patient was instructed about the treatment possibilities for occipital nerve neuralgia. After the suspicion of the diagnosis of ON, the patient underwent an anesthetic blockade in the region of the greater occipital nerve, with immediate relief of the symptoms, which corroborated the diagnosis. The patient underwent open-air rhizotomy of the greater occipital nerve with remission of symptoms for ~ 3 years, but with new symptoms present, which led her to undergo a new surgical intervention. Under general anesthesia, a skin incision was performed in the posterior midline, in the region between the external bulge and the spinous process of C7.[4] By means of careful dissection of the planes, the second spinal nerve was identified, ~ 3 cm from the midline (►**Fig. 1**). No tissue alterations were found in the proximal path of this spinal nerve, as well as no neuromas. A C2 neurectomy was performed bilaterally, proximal to the sensory ganglion (nerve extirpation, including the sensory ganglion). The patient was reevaluated four weeks after surgery, and showed improvement of the pain, without the aforementioned symptoms, only with discomfort in the cervical region and bilateral hypoesthesia in the occipital region. The patient did not present complications due to the surgical procedure during the first postoperative month. In the outpatient visits occurring in the third and sixth months, the patient presented improvement of the pain, returning to her work activities without any compromise.

Discussion

The greater occipital nerve originates from posterior branches of the segments of spinal nerve C2, after its emergence from the sensory ganglion.[4,5] It travels a recurrent path toward the lower edge of the lower oblique muscle of the head, circumventing it,

Fig. 1 Intraoperative photo of the occipital region. (**1**) External occipital bulge; (**2**) posterior tubercle of C1; (**3**) spinous process of C2; (**4**) second spinal nerve.

then travels in the upper direction, and, at this moment, is closely related to the semispinalis muscle of the head.[5] From this point on, it crosses the aforementioned muscle, maintaining its ascending path and establishing a new relationship when passing under the trapezius muscle. In its last portion, the nerve crosses the trapezius fibers, exteriorizing in the subcutaneous cell tissue.[5]

Occipital neuralgia is defined as pain in the occipital or cervical territory caused by stimulation and/or compression of the greater occipital nerve.[6] The pain has the characteristic of being unilateral and affecting the occipital region, which is innervated by the greater occipital nerve.[7] The patient can report it as pain in a burning or stabbing pattern, associated with the sensation of "shock," paresthesia, photophobia, nausea and vomiting.[8,9] Traumatic etiological, degenerative, oncologic or idiopathic factors are involved in most cases.[10]

The diagnosis of occipital neuralgia is essentially obtained by clinical examination, and no imaging exam presents good diagnosis efficiency.[11]

Different therapeutic approaches have been established for ON, which can be divided into conservative and surgical treatments. Among the options of conservative treatment are the use of cervical collar, analgesics, percutaneous nerve block, and use of botulinum toxin. As a non-ablative surgical treatment, neurostimulation of the greater occipital nerve is highlighted. Finally, in the ablative surgical treatment group are radiofrequency rhizotomy, Lissauer tract injury surgery, greater occipital nerve neurectomy, and neurectomy of spinal nerve C2.[2,12–14]

Conservative treatment of ON

The use of botulinum toxin has been shown to be efficient as therapy for ON, especially when associated with spinal cord

trauma.[8] It is noteworthy that it is already used in the treatment of other types of headache, such as migraine.[8] Infiltration of the greater occipital nerve can be used as diagnosis and treatment of this neuropathy. Infiltration can be performed with 0.5% bupivacaine, which may or may not be associated with methylprednisolone, in an anatomical point located 3 cm laterally and 2 cm inferiorly to the external occipital protuberance.[2,9] This nerve block showed positive results in the treatment of other types of headache. A hypothesis that justified good pain control is the proximity of the sensory neurons of the upper cervical region with neurons present in the trigeminal spinal nucleus.[11] Among the complications resulting from percutaneous nerve block due to its superficial location is intravascular injection.[1] In a study[2] involving 92 patients treated with percutaneous lidocaine and corticoid infiltration, good pain control was observed in 87% of the patients, with recurrence in 31.5% of them.[2]

Non-Ablative Surgical Treatment of ON

Neurostimulation in the greater occipital nerve has efficiency in the control of ON pain that ranges between 60% and 90%.[10] Among the complications of this surgical technique, we highlight the malposition of the electrodes and their migration, which can occur in 10% to 70% of the cases, with subsequent loss of stimulation and frequent indication of surgical revision.[10] A study[10] using an open technique for neurostimulation, despite the small sample, demonstrated that this technique is a safe, simple and effective modality to maintain direct contact between the electrode and the nerve trunk in a patient previously under anesthesia.[10] Medullary stimulation of C1-C4 has also been an option for the control of neuropathic pain in cases of ON.[15]

ON Ablative Surgical Treatment

In this treatment modality, open-pit rhizotomy of the greater occipital nerve is described for the treatment of ON.[5] Despite the small sample, one study[2] retrospectively evaluated 17 patients undergoing open-pit rhizotomy. No major complications were observed, but the potential risks are: infection, paralysis, cerebrospinal fluid fistula, and paresis of the trapezius muscle by the injury of the spinal accessory nerve.[2]

The surgery of the Lissauer tract injury to the C1-C3 dorsal roots has also been described as a treatment of ON. It is a modality of invasive surgical treatment, due to the need for cervical laminectomy. This surgery presents a higher complication rate when compared with other treatment modalities. Among the complications described are surgical wound infection, blood loss and spinal cord injury.[16]

Neurectomy of spinal nerve C2 is described as an option of ablative treatment of ON.[14] This technique has also been described in the arthrodesis of C1-C2, with the aim of minimizing pain or avoiding neuropathic pain postoperatively.[14] Among the reported complications are hypoesthesia and hyperesthesia of the occipital region.[14] It is important to emphasize that these described complications could be partly due to spinal cord trauma in this group of patients.[14]

Conclusion

Occipital neuralgia requires careful investigation to establish the diagnosis. Although there are several treatment methods, neurectomy of spinal nerve C2 may be an option for this pathology, having the advantage of being inexpensive. This technique requires anatomical knowledge for the proper recognition of the anatomical structures involved. Studies to better define the functional outcomes in the long-term are still necessary.

Conflict of Interests
The authors have no conflict of interests to declare.

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